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Antepartum hemorrhage from previous-cesarean-sectioned uterus as a potential sign of uterine artery pseudoaneurysm*

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Postpartum hemorrhage (PPH), a leading cause of maternal mortality, can occur within 24 h of delivery (primary PPH), or during the period from 24 h after delivery to Week 6 of puerperium (secondary PPH). It requires health professionals to be alert to the symptoms to ensure prompt diagnosis and treatment, especially in the case of rupture of a uterine artery pseudoaneurysm (UAP) due to its life-threatening consequence (Baba *et al*., 2014). Most of the published case reports or case serials describe UAP as a possible cause of delayed PPH after traumatic procedures during delivery or pregnancy termination, including cesarean section (CS), manual removal of the placenta, or dilation and curettage (D&C) (Wald, 2003). Herein, we report a case of prior CS-related UAP manifesting as primary PPH after an uncomplicated vaginal delivery. This case required emergency embolization and is notable for several reasons. Antepartum hemorrhage of the previously scarred uterus was a potential sign of the ruptured UAP, and color Doppler sonography sometimes deceived the physician as the characteristic features of UAP did not appear to be present.

A 37-year-old woman (gravid 2, para 1) at 38 weeks of gestation, visited our hospital with regular uterine contractions without obstetrical complication. The patient had undergone previous CS from delivering a healthy baby six years ago due to breech presentation. After discussion with the patient the chosen birth method was spontaneous vaginal delivery with close monitoring. Sudden vaginal fresh bleeding (100 ml) was observed. Placental abruption, vasa previa, and genital tract laceration were excluded as possible causes, and artificial rupture of membrane found clear amniotic fluid without trace of blood. However, the sudden fresh bleeding from a previous CS warranted further attention and informed consent procedures for an emergency CS were started.

An infant weighing 3190 g with Apgar scores of 10 at 1 min or 5 min was vaginally delivered 30 min later, followed by intact placenta delivered spontaneously. Total delivery time was 4 h with 250 ml blood loss within 2 h postpartum. The placenta showed no evidence of abruption.

This patient had massive vaginal bleeding accompanied by a large quantity of blood clots at 12 h postpartum. Physical examination indicated a well-contracted uterus, soft and non-distended abdomen, and mild suprapubic tenderness on deep palpation, and vaginal inspection revealed copious amounts of bright blood clot accumulating in the vaginal vault with no active bleeding from the cervical os or laceration of the genital tract. Intravenous oxytocin (20 units), intramuscular carboprost tromethamine, manual uterine massage, and packing the vaginal vault with multiple gauze were performed immediately. The vital signs of the patient were stable after giving crystalloid, packed red blood cells and fresh frozen plasma.

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However, the bleeding was persistent with total blood loss of 1800 ml within 40 min. A CS incision-related vascular problem was considered and could not be excluded with the assistance of color Doppler sonography. An interventional radiologist was called urgently for a pelvic digital subtraction angiography, revealing a large saccular aneurysm of the left uterine artery (Figs. 1a and 1b). Active bleeding was confirmed using contrast extravasated from the left uterine artery and dispersed into the cervical segment (Fig. 1b). Complete occlusion of the vessel was achieved following embolization with a regular size (Fig. 1c), and then a large size, gelatin sponge (Fig. 1d). The angiogram of the right uterine artery demonstrated an engorged and tortuous uterine artery (Fig. 1e), and a gelatin sponge was applied (Fig. 1f). This patient recovered smoothly, and was discharged on Day 4 postpartum. Specifically, a size 6-Fr Cobra catheter was inserted followed by 3–5 ml of contrast injection, and an absorbable gelatin sponge of 1 mm×1 mm×1 mm was used rather than a permanent embolization agent such as metallic coils or a glue suitable for greater vessels.

UAP has been considered a rare disorder. It is believed that pseudoaneurysm formation is due to a local traumatic procedure leading to vascular injury (Wald, 2003). Surgical or interventional procedures, such as CS or D&C, were considered the culprit in almost all reported cases of UAP manifesting as secondary PPH.

Controversial reports demonstrate that UAP also occurs after non-traumatic vaginal delivery (McGonegle *et al*., 2006). Since McGonegle *et al*. (2006) reported for the first time that UAP had occurred after uncomplicated spontaneous vaginal delivery in a patient without previous surgery, other

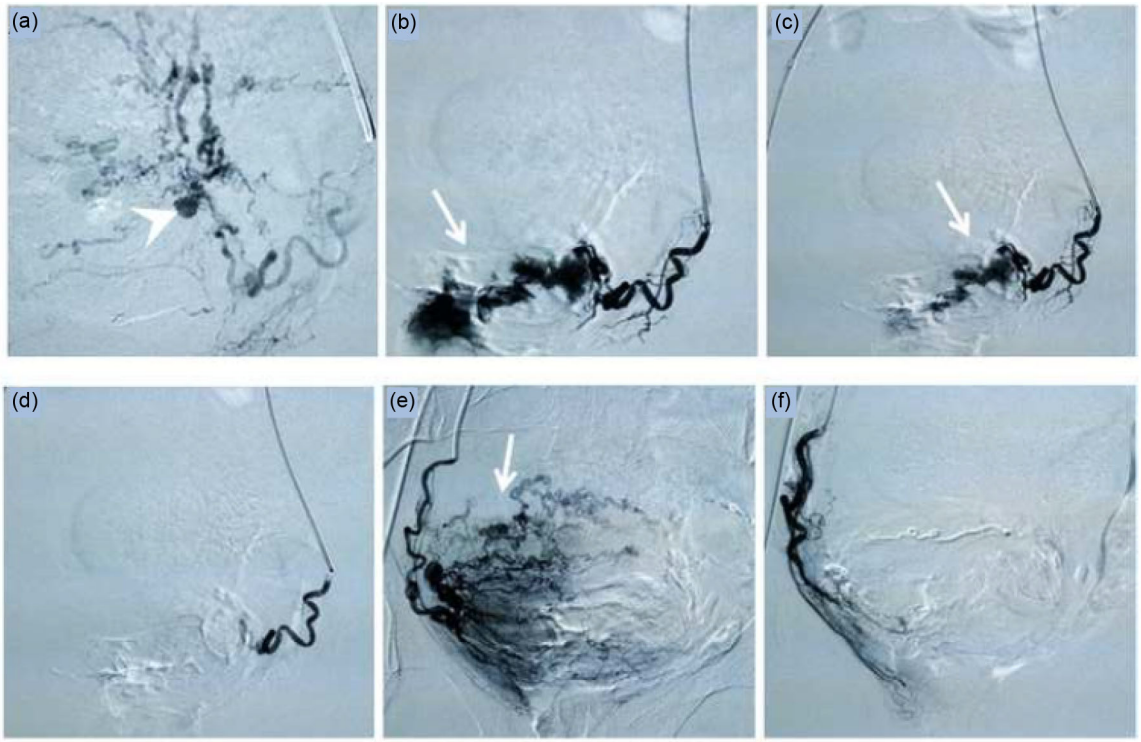

Fig. 1 Digital subtraction pelvis angiography and embolization of uterine artery pseudoaneurysm

(a) Digital subtraction angiogram of left uterine artery angiography demonstrates a pseudoaneurysm of the left uterine artery at the proximal portion (arrowhead). (b) Selective left uterine artery angiography shows an active extravasation (arrow) of contrast material into the uterine cavity and cervical segment, indicating active bleeding. (c) A selective left uterine artery angiogram obtained after the first embolization session demonstrates incomplete occlusion of target artery and continuous yet decreased extravasation (arrow) of contrast materials. (d) A selective left uterine artery angiogram obtained after enhanced embolization confirmed no further extravasation and complete exclusion of the left uterine artery. (e) A selective right uterine artery angiogram demonstrates an engorged and tortuous uterine artery (arrow), but pseudoaneurysm and extravasation were not identified. (f) A selective left uterine artery angiogram obtained after regular embolization shows decreased blood supply of the branch of left uterine artery

clinicians have subsequently reported similar scenarios (Baba *et al*., 2016). There is so far a total of 18 reported cases of UAP rupture after non-traumatic delivery/pregnancy termination (including our patient) in the literature, identified by a thorough PubMed literature search. Six (33.3%) of these were cases of primary PPH and 12 (66.7%) were secondary. The average interval between the preceding delivery/pregnancy and hemorrhage was 28 d.

The mechanism of UAP formation after non-traumatic procedure is still largely unknown due to the diversity of its clinical features. Underlying vascular abnormality, strong shear or acute stretching of the artery due to precipitous delivery, and infection-related damage to the adjacent artery wall have all been proposed as the potential mechanisms (Matsubara *et al*., 2014a). Retrospectively, there were several risk factors for UAP formation in our case including the prior CS, a relatively precipitous labor, and minor laceration of the cervix. Especially, it should be noted that antepartum hemorrhage was probably an early sign of the UAP rupture, and the precipitous labor followed by contraction of the uterus temporarily occluded the ruptured UAP, avoiding instant massive postpartum bleeding.

Prompt diagnosis is the key factor in dealing with the rupture of a UAP as this life-threatening emergency can lead to a devastating outcome in a very short space of time. Angiography and ultrasonography are non-invasive and clinically useful modalities for achieving a definitive diagnosis. However, under-diagnosis with bedside color Doppler sonography has been rarely reported (McGonegle *et al*., 2006; Gondo *et al*., 2014; Matsubara *et al*., 2014b; 2016). To our knowledge, there is no published figure for sonographic sensitivity in relation to UAP. The failure of color Doppler sonography to diagnose UAP in our case could be explained by the sensitivity of the sonography. Additionally, emergency transcatheter arterial embolization of the uterine arteries is a well-established technique (Soyer *et al*., 2008; 2015; Dohan *et al*., 2013) once rupture of UAP is highly suspected or diagnosed via the color Doppler sonography or computed tomography (CT) imaging. In our case, although the color Doppler did not confirm UAP, the risk factors involved in this patient as well as the intractable primary PPH unresponsive to conservative treatment still rendered the ruptured UAP in the priority list of differential diagnosis, and thus emergent digital subtraction angiography (DSA) was performed identifying the cause, and corresponding treatment with embolization was followed successfully.

In conclusion, our report further raises clinician awareness about the occurrence of ruptured UAP in the following ways. Firstly, UAP can occur even during an uncomplicated vaginal delivery, and may not necessarily be linked with "traumatic procedure-associated consequence". Secondly, antepartum hemorrhage of a previously scarred uterus may be a potential sign of ruptured UAP (Cornette *et al*., 2014), and further imaging tests are strongly recommended in suspected cases before the patient is discharged. Thirdly, urgent pelvic angiography may be essential in highly suspected UAP patients even if there is no conclusive evidence from color Doppler sonography due to its limitations with both sensitivity and/or experience.

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Compliance with ethics guidelines

Ning ZHANG, Wei-hua LOU, Xue-bin ZHANG, Jian-hua LIN, and Wen DI declare that they have no conflict of interest.

All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1975, as revised in 2008 (5). Informed consent was obtained from the patient for being included in the study. Additional informed consent was obtained from the patient for which identifying information is included in this article.

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中文概要

题　目：疤痕子宫产前出血可能是子宫动脉瘤的潜在危险信号

目　的：子宫动脉瘤多被认为与产程中创伤性操作相关，但近年陆续有报道显示子宫动脉瘤可发生于自然分娩过程中。本文将为自然分娩过程中子宫动脉瘤的发生和紧急处理等提供参考指导。

创新点：产前出血可能是子宫动脉瘤发生的潜在危险信号。

方　法：病例报道和文献汇纳分析。患者女，37 岁，疤痕子宫，临产入院，产前出血，经阴顺产后 12 h，突发阴道大量出血，常规对症处理（按摩子宫、阴道填塞、缩宫素静滴和欣母佩宫颈注射等）未能有效缓解，40 min 内出血达 1800 ml。床边 B 超未提示动脉瘤特征，数字减影血管造影（DSA）显示左侧子宫动脉瘤，即刻予动脉栓塞成功止血。患者产后 4 d 无并发症出院，随访无后遗症。

结　论：临床医师需要加强对子宫动脉瘤发生的警惕认识和急救处理水平。子宫动脉瘤和产程中的有创性操作无必然相关性，疤痕子宫的产前出血可能是子宫动脉瘤发生的潜在危险信号，产后随访应提高对晚期出血发生的警惕性。鉴于 B 超在诊断动脉瘤方面的有限性，高度怀疑动脉瘤时应尽快行血管造影明确诊断，及时栓塞处理。

关键词：子宫动脉瘤；经阴分娩；早期产后出血；动脉栓塞